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A case of primary malignant fibrous histiocytoma of the duodenum

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ABSTRACT

Background: Primary malignant fibrous histiocytoma (MFH) of the duodenum is rare and a distinct clinical entity.
Case report: A 55-year-old man presented with a history of upper gastrointestinal bleeding, vomiting and decreased body weight over the past 2 months. Abdominal exam showed an epigastric mass of 10 cm. An upper gastrointestinal endoscopy documented a tumor in the third part of duodenum. The histopathological examination of biopsy has concluded a MFH. Abdominal CT revealed a large and heterogeneous mass of 10 cm in the third part of the duodenum. The intervention was conducted by way of a bi-subcostal laparotomy. Exploration of the tumor revealed involvement of the third part of duodenum. This lesion adhered and invaded the inferior vena cava. A palliative procedure using a gastro-entero-anastomosis was carried out with uneventful postoperative course. Neither adjuvant chemotherapy nor radiotherapy were conducted. The patient died four months following his operation.
Conclusion: The biological behavior of malignat fibrous histiocytomas is extremely aggressive and mainly conditioned by size and histological grading. The treatment of choice, whenever possible, is based on early and complete surgical excision of the tumor.

1. Introduction

Malignant fibrous histiocytoma (MFH) represents the most common soft tissue sarcoma among adults.[1–3] Visceral locations are uncommon and primary duodenal MFH is exceedingly rare.

2. Observation

We present a case of a 63-year-old-man who was admitted to our hospital because of upper and highly abundant gastrointestinal bleeding (Hg = 4 g/dl), epigastric mass of 10 cm, vomiting, lower extremities edema and weight loss. After resuscitation and stabilization of the patient (Hg = 11 g/dl), an upper gastrointestinal endoscopy was performed that showed a large tumor of the third part of the duodenum, which was completely stenosed. Histologic examination of endoscopic biopsies of the tumor showed proliferation of both spindle (fibroblast-like) and round (histiocyte-like) cells, arranged in a cartwheel-like (storiform) and accompanied by pleomorphic giant cells and inflammatory cells. The tumor was then classified as a storiform–pleomorphic type malignant fibrous histiocytoma (Fig. 1) according to the classification suggested by Weiss and Enzinger.[1] Abdominal CT scan (Fig. 2) indicated a large and heterogeneous mass of 15 cm in the third part of the duodenum. Neither locoregional nor distant metastatic invasions were detected.

The intervention was conducted by way of a bi-subcostal laparotomy. Exploration of the tumor revealed involvement of the third part of the duodenum (Fig. 3). Moreover, solid and cystic lesions were observed with the help of an intraoperative ultrasound examination. These lesions adhered and invaded the inferior vena cava. A palliative procedure using a gastro-entero-anastomosis was carried out with uneventful postoperative course. Neither adjuvant chemotherapy nor radiotherapy were conducted. The patient died four months following his operation.

3. Discussion

Since the first report of MFH in 1964 by O'Brien and Stout, MFH has been well recognized as the most common malignant soft tissue tumor. It usually occurs in the deep planes of proximal extremities, in the retroperitoneum and in the trunk, yet, it can occur almost anywhere because of its mesenchymal origin, including bones.[4] Rarely, this tumor is primarily encountered in organs such as the brain,[5] lung,[6] heart,[7] breast,[8] pancreas,[9] gallbladder,[10] or the alimentary tract.[11] To date, and to the best of our knowledge, only 6 cases of malignant fibrous histiocytoma of the duodenum have been reported. Five of them were primary MHF including this present case.[12–15] Only one case of metastatic MFH to the duode-

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Table 1
Up-to-date review of cases of primary malignant fibrous histiocytoma of the duodenum.

Case n	Source	Age (sex)	Symptoms	Size (mm) (Invasion, metastasis)	Therapy	Histologic type	Follow up
1	Gilman et al (12) 1986	29 (female)	Peritonitis	70 (None)	Pancreatoduodenectomy	Storiform-pleomorphic	Few days (died)
2	Asai et al (13) 1987	61 (male)	Epigastralgia	70 (None)	Resection	Pleomorphic	21 months (alive)
3	Farinon et al (14) 1999	61 (female)	Gastrointestinal bleeding	90 (None)	Pancreatoduodenectomy	Storiform-Pleomorphic	2 months (died)
4	Wang et al (15) 2005	61 (male)	Gastrointestinal bleeding, weight loss	150 (None)	Pancreatoduodenectomy	Storiform	24 months (alive)
5	Present case	63 (male)	Gastrointestinal bleeding, weight loss, vomiting	150 (Inferior vena cava)	gastroenteroanastomosis	Storiform-pleomorph	4 months (died)

num was described by Yarze and D'Agostino, and that occurred two years after surgical excision of the right anterior deltoid MFH.[16] We reviewed and summarized the findings from reported cases of primary duodenal MFH as indicated in the English literature, as well as presenting the findings from our case (see Table 1).

The mean age of the patients was 55 years, with a range of 29–63 years. The ratio of men to women was 1.5. Only one case (1/5, 20%) had local invasion (i.e. inferior vena cava) and none had a distance metastasis.

The common clinical symptoms were by order of frequency: gastrointestinal bleeding (3/5), abdominal pain (2/5) and weight loss (2/5). No specific complaints or findings at physical examination were known to distinguish primary MFH from other malignant tumors in the gastrointestinal tract. Therefore, the final diagnosis was made only after pathological and immunopathological examination of the tumor.

The tumor diameter ranged from 70 mm to 150 mm. In our patient, the mass measured 150 mm in size. He died after 4 months.

There are four main morphologic variants of MFH: storiform–pleomorphic, myxoid, inflammatory, and angiomatoid.[17] The storiform–pleomorphic pattern (5/5, 100%) of duodenal MFH was the only variant.

From reports found in the literature, there is still no consensus on the value of adjuvant or neoadjuvant chemo or radiation therapy in primary MFH of the duodenum. Conventional chemotherapy for MFH of the extremities has been reported to have little effect in prolonging the overall survival of MFH cases with small bowel.[18–20] Early and complete surgical excision may be the only form of management, which may provide the patient with good results. Our patient received a palliative procedure because of a local

Fig. 1. Histopathological examination. Above: proliferation of histiocyte-like elements, occasionally multinucleated with large nuclei and nucleoli, and fibroblast-like spindle cells, focally arranged in a storiform pattern (HE, 40×). Below: selected area from the foregoing picture displays high mitotic activity with numerous abnormal forms (HE, 200×).

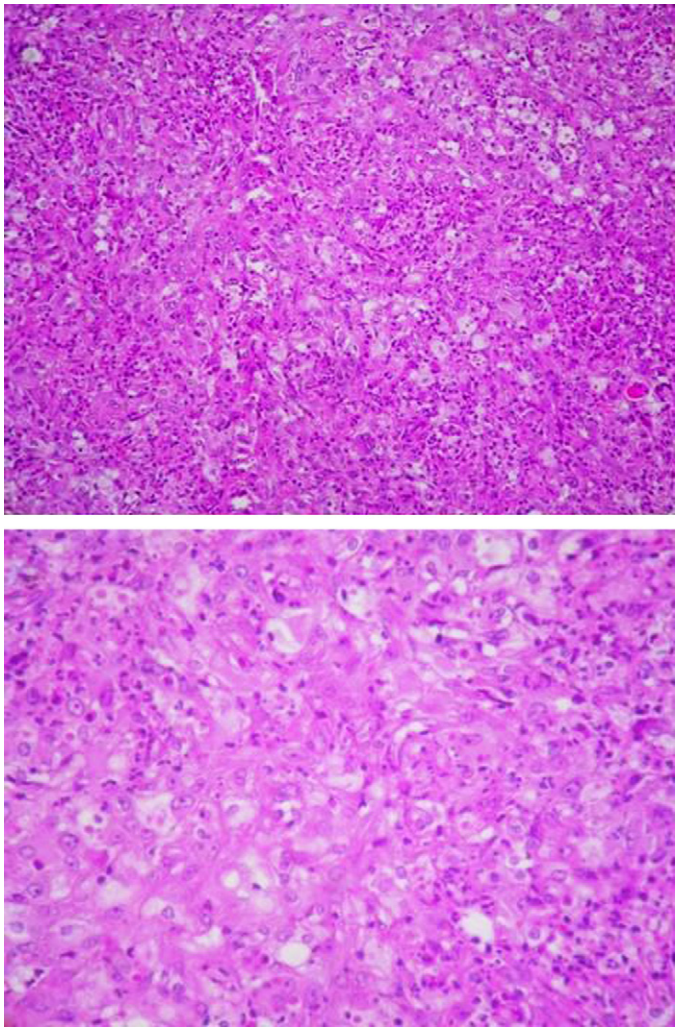

Fig. 2. Abdominal computed tomograph scan demonstrated a heterogeneous mass in the third part of duodenum (arrow). This mass contained a central necrosis.

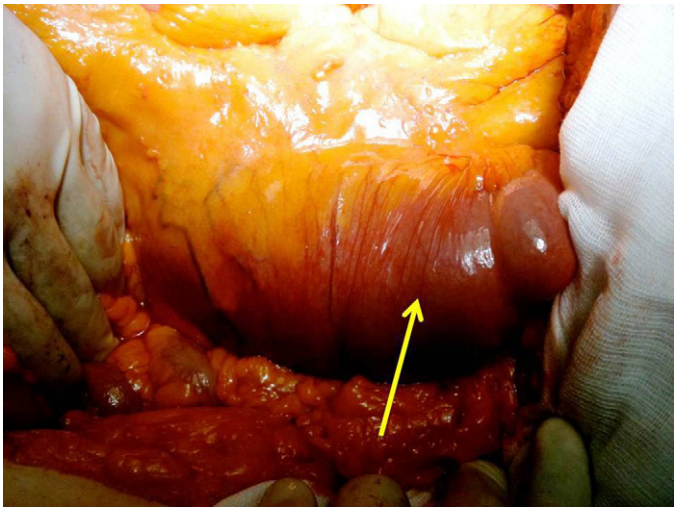

Fig. 3. Operative photograph showing a large tumor developed within a third part of duodenum.

invasion. Concerning prognostic factors, the site of the tumor represents undoubtedly an important prognostic factor for MFH located in the retroperitoneum or abdomen. Late detection of these tumors due to their location is responsible for a poorer prognosis, as compared with that of soft tissue tumors of the extremities. Weiss and Enzinger[1] found that larger and more deeply located tumors metastasized more frequently, but size was not as reliable as the depth of the tumor when correlated with metastatic potential.

4. Conclusion

In conclusion, while occurring rarely, MFH should be considered in the differential diagnosis of large duodenal lesions. Tumor size, tumor site, adjacent organ invasion or metastases at diagnosis remain the most important prognostic factor. The final diagnosis is made only after pathological examination of the tumor. Storiform–pleomorphic pattern in histology is the most common variant. When it is possible, the management of primary MFH of the duodenum should be surgery.

Conflicts of interest statement

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Ethical approval

Obtained.

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